Parents' preferences for the organisation of long-term follow-up of childhood cancer survivors

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Abstract: Parents take an important role in follow-up of young cancer survivors. We aimed to investigate (1) parents' preferences for organisation of follow-up (including content, specialists involved and models of care), and (2) parents' and children's characteristics predicting preference for generalist vs. specialist-led follow-up. We sent a questionnaire to parents of childhood cancer survivors aged 11-17 years. We assessed on a 4-point Likert scale (1-4), parents' preferences for organisation of long-term follow-up. Proposed models were: telephone/questionnaire, general practitioner (GP) (both categorised as generalist for regression analysis); and paediatric oncologist, medical oncologist or multidisciplinary team (MDT) (categorised as specialists). Of 284 contacted parents, 189 responded (67%). Parents welcomed if visits included checking for cancer recurrence (mean = 3.89), late effects screening (mean = 3.79), taking patients seriously (mean = 3.86) and competent staff (mean = 3.85). The preferred specialists were paediatric oncologists (mean = 3.73). Parents valued the paediatric oncologist model of care (mean = 3.49) and the MDT model (mean = 3.14) highest. Parents of children not attending clinic-based follow-up (OR = 2.97, p = .009) and those visiting a generalist (OR = 4.23, p = .007) favoured the generalist-led model. Many parents preferred a clinic-based model of follow-up by paediatric oncologists or a MDT. However, parents also valued the follow-up care model according to which their child is followed up.

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Parents' preferences for the organization of long-term follow-up of childhood cancer survivors

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Abstract
Parents take an important role in follow-up of young cancer survivors. We aimed to investigate 1) parents' preferences for organization of follow-up (including content, specialists involved and models of care), and 2) parents' and children's characteristics predicting preference for generalist versus specialist-led follow-up. We sent a questionnaire to parents of childhood cancer survivors aged 11-17years. We assessed on a four-point Likert scale (1-4), parents' preferences for organization of long-term follow-up. Proposed models were: telephone/questionnaire, general practitioner (GP) (both categorized as generalist for regression analysis); and pediatric oncologist, medical oncologist or multidisciplinary team (MDT) (categorized as specialists). Of 284 contacted parents 189 responded (67%). Parents welcomed if visits included checking for cancer recurrence (mean=3.89), late effects screening (mean=3.79), taking patients seriously (mean=3.86, SD=0.35) and competent staff (mean=3.85). The preferred specialists were pediatric oncologists (mean=3.73). Parents valued the pediatric oncologist model of care (mean=3.49) and the MDT model (mean=3.14) highest. Parents of children not attending clinic-based follow-up (OR=2.97, p=0.009) and those visiting a generalist (OR=4.23, p=0.007) favored the generalist-led model. Many parents preferred a clinic-based model of follow-up by pediatric oncologists or a multidisciplinary team. However, parents also valued the follow-up care model according to which their child is followed up.

Key words: parents of childhood cancer survivors; pediatric oncology; follow-up care; models of care; cancer registry, Europe

INTRODUCTION

Lifelong follow-up care is recommended for most childhood cancer survivors. The goal of follow-up is to identify and treat relapse and late effects early, and provide age-adapted information about cancer, treatment, potential late effects and health behavior. (Bhatia & Meadows, 2006; Hudson , et al., 2013; Oeffinger , et al., 2006; Taylor, Absolom, Snowden, Eiser, & Late Effects Group, 2012). Guidelines have been developed to provide recommendations for risk-stratified long-term follow-up care (Children's Oncology Group, 2008; Scottish Intercollegiate Guidelines Network (SIGN), 2004; Wallace, Thompson, Anderson, & Guideline Development, 2013). Various models of care have been described and compared (Heirs , et al., 2013) such as follow-up by telephone (James, Guerrero, & Brada, 1994), multidisciplinary team (MDT) (Carlson, Hobbie, Brogna, & Ginsberg, 2008; Edgar & Wallace, 2008), pediatric oncologist (Haddy & Haddy, 2010), general practitioners (GP) (Schmidt , et al., 2010) or shared-care models (Blaauwbroek, Tuinier, Meyboom-de Jong, Kamps, & Postma, 2008). In many countries, none of the models have been implemented and long-term follow-up is not standardized.

In Switzerland, childhood cancers survivors are regularly followed-up by their pediatric oncologist into their early twenties, and are then usually discharged to a GP or medical oncologist. Others may continue follow-up with their pediatric oncologist longer into adulthood.

Prior to setting up a specific model of follow-up care survivors' and their parents' opinions and preferences for the organization of care should be assessed (Aslett, Levitt, Richardson, & Gibson, 2007; Earle, Davies, Greenfield, Ross, & Eiser, 2005). Survivors' opinions and preferences have previously been studied (Michel , et al., 2016; Michel , et al., 2009). Parents' expectations of follow-up care have only been addressed in a small focus group study in the UK (Earle , et al., 2005): parents desired medical facts and written test results for reassurance, information on psychosocial consequences, and wanted to have the possibility to meet other families with a child survivor. Parents did not value the GP model since they perceived that specialist knowledge was not available.

Parents take an important role in follow-up care for many reasons: they are most aware about the child's medical history but also provide things such as transportation or reminding about doctor's appointment. We thus aimed to investigate 1) parents' preferences for the organization of follow-up care (including content, specialists involved and different models of care). These outcomes were assessed for both children attending and not attending clinic-based follow-up. 2) We investigated associations of socio-demographic characteristics of parents and children's clinical factors with preferences for generalist versus specialist-led follow-up.

METHODS

Sample and procedure

The Swiss Childhood Cancer Registry (SCCR) is a national population-based cancer registry including all cancer patients, diagnosed with leukemia, lymphoma, central nervous system (CNS) tumor, malignant solid tumor or Langerhans cell histiocytosis at age <21 years in Switzerland since 1976 (Michel , et al., 2007; Michel , et al., 2008). The Swiss Childhood Cancer Survivor Study (SCCSS) is a nationwide, long-term follow-up survey of the SCCR including a baseline (2007-2011) and a follow-up questionnaire (2010-2012).

The baseline questionnaire included all patients registered in the SCCR who were diagnosed between 1976-2005, aged <21 years and survived for ≥5 years (Kuehni , et al., 2012a). Parents of survivors aged ≤15 years completed the questionnaire for their children, whereas survivors 16+ years completed their own questionnaire.

The follow-up questionnaire was sent approximately 2 years later. Parents who filled in the baseline questionnaire were contacted again if their child who had cancer was aged 11-17 years at time of follow-up study (eligible N=306; **Supplemental Figure 1**). They received the questionnaire with a prepaid return envelope, and if they did not reply within two months, a reminder letter with another questionnaire. Questionnaires were available in German and French and focused on topics related to follow-up care.

Ethics approval was provided through the general cancer registry permission of the SCCR (The Swiss Federal Commission of Experts for Professional Secrecy in Medical Research) and a non-obstat statement from the ethics committee of the canton of Bern declaring that the ethics committee did not object the conduct of the study.

Measurements

The follow-up survey of the SCCSS focused on follow-up care and psychological outcomes. Follow-up care after childhood cancer was introduced on the front page of the questionnaire as follow-up appointments of their child due to the previous severe disease.

Outcome

Items were purpose designed and based on a previous study in the UK (Michel , et al., 2009).

What are the reasons for follow-up: Parents rated the importance (1="not at all important" to 4="very important"; this scale was used in all questions where parents had to rate importance) of different reasons for attending follow-up (nine items).

What should be included in follow-up: We asked parents about the perceived importance of four medical aspects and eight general aspects of follow-up (**Figure 1A**).

What is important during appointments: Parents rated the importance of 10 organizational aspects (**Figure 1A**).

Who should be involved in follow-up: Parents rated the importance of different medical and other specialists involved in follow-up (**Figure 1B**).

Models of care: We provided a short description of five different models of follow-up care by: a) telephone/questionnaire, (led by a nurse referring patients to specialist care if needed), b) GP-led and referring patients to a specialist if needed, c) pediatric oncologist who originally treated the survivor, d) medical oncologist, e) multidisciplinary team (MDT) in a hospital (defined as including several specialists such as oncologists, endocrinologists, psychologists, social workers and nurses, all of whom are accessible during one appointment). For each model we asked parents' agreement to four items (1="don't agree at all" to 4="completely agree"): 'it would suit my child', 'I am afraid that health problems are not detected', 'I am not satisfied with this kind of follow-up', this model of follow-up is appropriate for the needs of their child.

Explanatory variables assessed by questionnaire

We assessed parents' sex, age at study, migration background (migration if they were not Swiss citizens since birth or not born in Switzerland), language region (German vs. French), parents' employment status (employed vs. not employed), and education (three categories: primary (compulsory schooling including vocational training/apprenticeship); secondary (teachers/technical and commercial schools etc.); tertiary (university and university of applied sciences; **Table 1**) (Kuehni , et al., 2012b).

Additionally, we asked parents if their child still attended follow-up: 1) 'yes, my child still attends regular follow-up appointments'; 2) 'yes, my child still has irregular follow-up appointments'; 3) 'no, regular follow-up is completed, but my child goes to the doctor for any cancer-associated complications'; 4) 'no, regular follow-up is completed and my child has not seen the doctor

for a while'. A binary variable was created: attenders (responses 1 or 2) and non-attenders (responses 3 or 4). Parents indicated on a list which doctors were involved in current care. This was coded as "specialist care" if parents listed at least one specialist and "generalist" if only a GP was indicated. Parents were asked whether they are currently involved in follow-up care (parental involvement=yes/no) (Vetsch , et al., 2016). Concerns of parents about consequences of their child's illness were assessed by the question "How concerned are you about consequences of your child's illness?" (adapted from the Brief Illness Perception Questionnaire (IPQ) using a 0-to-10 response scale) (Broadbent, Petrie, Main, & Weinman, 2006). The response was divided in three categories (no: 0-2, medium: 3-6, and high concerns: 7-10).

From the baseline questionnaire of the SCCSS we extracted information about parent-reported late effects of the survivor (yes/no) (Kuehni , et al., 2012a).

Clinical variables of the child extracted from the SCCR

We extracted medical information on cancer diagnosis and treatment of the child from the SCCR. Cancer diagnosis was classified according to the International Classification of Childhood Cancer (third edition) (Steliarova-Foucher, Stiller, Lacour, & Kaatsch, 2005). For the analyses we recoded diagnosis into six major groups: leukemia, lymphoma, CNS tumors, neuroblastoma, bone tumor/soft tissue sarcoma (STS) and other tumors. Treatment was coded as: surgery only, chemotherapy (without radiotherapy ±surgery), radiotherapy (±surgery and/or chemotherapy) and stem cell transplantation (SCT; may have had surgery and/or chemotherapy and/or radiotherapy). The type of treating hospital was divided into university and regional hospital. Age at diagnosis was divided into three categories: 0-1 year, 2-4 years, ≥ 5 years. We have chosen these categories because they might influence risk for late effects and preferences for follow-up care. Age at study was divided into three categories: <14 years, 14-15 years, >15 years. Time since diagnosis was divided into three categories 5-9 years, 10-14 years and 15-17 years. Relapse was coded yes/no.

Analyses

Analyses were performed using Stata 13.1 (StataCorp, College Station, TX).

We used descriptive statistics to compare participants and non-participants of the study. For aim 1 we calculated means of the responses for each item. Paired t-tests were used to compare the means of clinical and supportive reasons. Hotelling t-test was used for the comparison of more than two means. Principal component analysis was used to test individual item loading onto different factors. The preference-score for each model of care was calculated as the overall mean of the four items assessed for each model. Two items had to be reverse coded such that higher scores indicated higher agreement (1-4). For each parent we determined the model with the highest preference-score. We then calculated the proportion of parents indicating each respective model as their preferred one (Figure 2) and stratified it by the model their child is currently attending (Figure 3). Parents could have more than one model reaching the highest preference-score.

To analyze difference in preferences between attenders and non-attenders we used t-test and chi^2-test. Bonferroni correction was used to correct for multiple testing.

For aim 2 we determined the model with the highest mean for each parent and created a binary variable indicating if GP or telephone/questionnaire follow-up (generalist follow-up=1) or any other follow-up model was rated highest (pediatric, medical oncologist, MDT: specialist follow-up=0, Table 2). Telephone/questionnaire and GP led model were grouped into generalist model because survivors would first contact a health care provider not necessarily specialized in pediatric oncology and only be referred to a specialist if needed. We used univariable logistic regression to investigate associations of parents' and their child's characteristics with the preferences for generalist versus specialist follow-up.

RESULTS

Of 306 eligible parents, we traced and contacted 284 (**Supplemental Figure 2**). Of those contacted, 189 (67%) responded. The mean age of parents was 46.1 years (SD=4.8, range 33.5-59.5 years), mean age of the child at study was 14.8 years (SD=1.8, range 10.7-18.0 years), mean age at diagnosis was 3.4 years (SD 2.5 range 0-9.2 years) and the mean time since diagnosis 11.3 years (SD 2.5, range 6.8-17.2 years; **Table 1**). Most children were diagnosed with leukemia (39.2%) followed by CNS tumors (18.0%). Participating and non-participating parents were similar in socio-demographic and clinical characteristics (**Table 1**).

1) Parents' preferences for organization of follow-up care

What are the reasons for follow-up: Factor analysis revealed two scales: supportive care (get reassurance about health, talk to staff who understand my child has been through, get advice about how to stay healthy, receive psychological support, get advice about everyday things) and clinical care (get information about late effects, check the cancer has not come back, help clinic staff learn more about late effects, get the best medical care). Cronbach's alpha, a measure for internal consistency, was good for supportive care: α=0.73; but low for clinical care α=0.58. Parents valued clinical reasons (mean=3.75, SD=0.33) higher than supportive reasons (mean=3.11, SD=0.58; p<0.001).

What should be included in follow-up: Among clinical aspects, parents rated *check for cancer recurrence* as most important (mean=3.91, SD=0.36; **Figure 1A**), before *screen for late effects* (mean=3.79, SD=0.45; p<0.001) and *information on potential late effects* (mean=3.65, SD=0.57; p<0.001). Regarding general aspects, knowing about *risks for their child's offspring* was rated as most important (mean=3.12, SD=0.82) whereas *exchange with other former patients* (mean=2.29, SD=0.91) and *religion/spirituality* were rated least important (mean=1.68, SD=0.79).

What is important during appointments: Parents rated *patient is taken seriously* (mean=3.86, SD=0.35; **Figure 1A**) and *competent staff* (mean=3.85, SD=0.37) as most important aspects, and significantly more important than the *quality of relationship to medical staff* (mean=3.75, SD=0.52; p=0.002). Least important were *short consultation* (mean=2.55, SD=0.81) and *meet former patients* (mean=2.16, SD=0.82).

Who should be involved in follow-up: When we asked about staff who should be involved in follow-up care parents rated the *pediatric oncologist* as most important (mean=3.73, SD=0.68) and significantly more important than *general practitioners* (mean=3.28, SD=0.89; p<0.001; **Figure 1B**). Radiotherapist (mean=2.02, SD=0.95) and social workers (mean=1.95, SD=0.90) were least important.

Models of follow-up care: For each model, we calculated the overall mean score among all parents. Additionally, we calculated the number of parents who had the highest preference-score for the respective model compared to all other models (**Figure 2**). Most parents preferred follow-up by a pediatric oncologist (N=117, 61.9%, mean=3.49, SD=0.65), followed by MDT (N=72, 38.1%, mean=3.16, SD=0.74; p<0.001), GP (N=55, 29.1%, mean=2.71, SD=0.97) and medical oncologist (N=54; 28.6%, mean=2.84, SD=0.86). Only few parents preferred the telephone/questionnaire model (N=9, 4.8%, mean=1.81, SD=0.79). The pediatric oncologist and MDT model were rated significantly higher than the other three models (all p<0.001). When stratified for the model their child is currently attending, parents whose child attended specialist follow-up preferred the pediatric oncologist model (44.6%) followed by MDT (21.6%). Parents whose child saw a generalist preferred the GP model (29.1%) but also had preferences for specialist-led follow-up care (**Figure 3**).

There was no difference in preferences for follow-up between parents of attenders and non-attenders after Bonferroni correction (**Supplemental Table 1)**.

2) Associations with parents' preferences for generalist versus specialist-led follow-up:

We used logistic regression analyses to determine characteristics of parents and clinical characteristics of the child associated with preferences for generalists follow-up (GP and telephone/questionnaire) versus specialist follow-up (pediatric or medical oncologist, MDT; **Table 2**). Parents of children not attending follow-up care (OR=2.97, CI 1.33-6.60, p=0.009) or already visiting a generalist for follow-up (OR=4.23, CI 1.84-9.71, p=0.007) rated the generalist model higher. A trend could be seen for lower preferences for generalist follow-up care for parents of children who had had a relapse (OR=0.23, CI 0.03-1.78, p=0.083) and who had been treated in a regional hospital (OR=0.31, CI 0.07-1.39, p=0.080).

DISCUSSION

We found that clinical reasons to attend follow-up were more important than supportive reasons to parents of childhood cancer survivors aged 11-17 years. Medical aspects such as checking for cancer recurrence or screening for late effects were rated as most important. Parents wanted that their child is taken seriously and competent staff is available. Pediatric oncologists and GPs were rated as the preferred doctors. Parents' preferred model of care was pediatric oncologist-led follow-up or follow-up by a MDT. The generalist model was only favored by parents of children not attending follow-up care at a treating clinic or who already see a generalist.

The importance of medical aspects during follow-up was already reported in previous studies (Christen , et al., 2016; Earle , et al., 2005; Eiser, Levitt, Leiper, Havermans, & Donovan, 1996; Michel , et al., 2016; Michel , et al., 2009). We reported that screening for late effects and check for cancer recurrence was rated most important for parents which are in line with what survivors reported. Parents want to be reassured about the cancer and know that their child is in best current health. A focus group analysis of parents of survivors aged 13-25 years showed that it is important to learn about risks for future health but also about how to stay healthy (Earle , et al., 2005). This is in contrast to our results where the general aspects such as risk for offspring were rated less important. The survivors in our samples are young and future health might not be of biggest concern, however parents and survivors should be informed that many years after diagnosis the risk for cancer recurrence diminishes and follow-up care is of higher importance to screen for late effects and learn about healthy lifestyle (Reulen , et al.).

When we asked for the specialist which should be involved parents preferred the pediatric oncologist followed by the GP. This preference for the pediatric oncologist was in line with another Swiss study on childhood cancer survivors (Michel , et al., 2016). In contrast to our findings a previous focus group reported that follow-up at a GP was evaluated as not appropriate since the specialist knowledge was lacking, whereas clinics led by specialist nurses were perceived as more acceptable, in offering both specialist expertise and opportunities for appropriate feedback (Earle , et al., 2005). However, these survivors were still in clinic-based follow-up most likely by a pediatric oncologist and therefore possibly favoring this specialist. Even though second highest in our study, another study in the US on survivors showed that follow-up by primary care physicians was rated highest and the late effects specialist second highest only (Zebrack , et al., 2004). Concerning might be that other studies reported that generalists lack knowledge and information on potential late effects or comfort of care for childhood cancer survivors (Lawrence, McLoone, Wakefield, & Cohn, 2016; Mertens , et al., 2004). Therefore, a close collaboration with specialist should be guaranteed and educational interventions for GPs if required organized. A Dutch study showed that GP are willing to follow-up childhood cancer survivors in a shared-care model, however they saw lack of information and communication as a barrier (Blaauwbroek , et al., 2007). Therefore written treatment summaries or a passport for care should be provided and help the GP guide through recommended screening and follow-up care processes (Horowitz, Fordis, Krause, McKellar, & Poplack, 2009). Such a passport will be implemented across Switzerland within the next years.

We additionally showed that most parents preferred follow-up care by a pediatric oncologist or a MDT led model. The telephone/questionnaire led model was least preferred. This is in line with two other studies among survivors who reported the pediatric oncologist-led follow-up as most important and the telephone/questionnaire follow-up least important (Michel , et al., 2016; Michel , et al., 2009). However, in the UK they only included survivors who attended clinic-based follow-up which was most likely led by a pediatric oncologist. Also expert committees have often favored long-term follow-up care clinics led by a MDT because late effects might be diverse and complex (Wallace , et al., 2001). With the ever growing population of survivors, follow-up care by pediatric oncologists however will not be feasible and manageable in Switzerland and MDT models might be too cost intensive. Therefore, in Switzerland many survivors are transitioned to a GP. Our results also indicated that parents of Swiss survivors still seeing a pediatric oncologist favor the pediatric oncologist-led follow-up, and parents whose children see a GP favor the generalist model. These parents also know specialist care from the first 5-10 years follow-up by the pediatric oncologist. Our results thus suggest that a risk-stratified approach where low-risk survivors are transferred to GP-led follow-up could meet parents' preferences. These findings are supported by another Swiss study where we showed that adolescent and young adult survivors preferred follow-up by medical oncologists, most of whom were treated by medical oncologists (Christen , et al., 2016). As shown in another study on adult survivors satisfaction with care did not depend on the clinic type but rather on shorter waiting time and possibilities to discuss health concerns (Absolom , et al., 2006). Parents and survivors preferences and satisfaction of care should be taken into account as it might ensure future attendance in follow-up.

We found no other clinical or socio-demographic associations for preferences for generalist-led follow-up care. Neither diagnosis nor late effects were associated with different preferences for follow-up care. However, there was some indication for a generalist preference in parents of survivors who did not have a relapse and those treated at a university hospital. This might suggest that preferences do not reflect the risk for late effects.

Our results and previous findings suggest that survivors and parents might be happy and feel comfortable with the model of care their children are currently receiving. Preference of care might be related to the satisfaction of care even though not measured in our study. Parents' preferences of care should therefore be considered early on and, if possible, follow-up care should be framed taking their preferences into account. Being the primary caregiver of young survivors their preference and satisfaction of care might ensure later attendance at follow-up care. However, parents should also be given adequate assurance and support in taking the decision on the future health care provider. Alternative models and individual preferences of long-term follow-up should be discussed. Additionally, both health care providers and primary caregivers might profit from written treatment summaries and survivorship care plans and guarantee adequate follow-up. A risk-stratified approach, where survivors receive follow-up care depending on diagnosis, and treatment (indicating their risk for late effects) might be the most adequate approach (Eiser , et al., 2006). However, for Switzerland such an approach has so far not been implemented (Rebholz , et al., 2011).

A limitation of this study is self-selection: parents of specific groups such as parents with greater interest in follow-up care or with higher needs may have been more willing to complete the questionnaire, others have been excluded because they did not complete the baseline questionnaire. Additionally, we only contacted one parent, mostly mothers, and thus information on preferences of the other parent is lacking. Also, we did not contact the survivors themselves in these families, and thus the preference of care of survivors is lacking. Another limitation is that we cannot tell if this is what parents really prefer or what they have been told to do by the treating physician. Further, we were not able to stratify survivors according to their risk because detailed information on exact treatment was lacking. The small sample size resulted in reduced precision and large confidence intervals. Therefore, only limited stratification of results was possible. Other limitations are the low reliability of the scale "clinical reasons" and the self-reported late effects.

Despite the relatively small sample size, this is a study with a rather large sample of parents of childhood cancer survivors compared to previous research. We were able to include parents of survivors attending and not attending clinic-based follow-up, and included prospectively collected data from the SCCR and from two questionnaires from the SCCSS. The response rate was good (67%).

Follow-up is an important aspect of quality of survivorship. In the transitioning phase from child to adult care it is important to not only meet survivors' or providers' preferences, but also parents' preferences for the organization of follow-up care. This might avoid a future loss to follow-up. We showed that many parents prefer a clinic-based model of follow-up by pediatric oncologists or a multidisciplinary team. However, parents also valued the follow-up care model according to which their child is followed up.

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Table 1. Characteristics of the study population, comparing parents participating and not participating in the questionnaire survey

	Participating parents		Non-participating parents	
	N	%	N	%
Total	**189**	**100**	**117**	**100**
Socio-demographic characteristics of parents				
Sex				
Female	160	84.7	n.a.[d]	
Male	29	15.3	n.a.	
Age at study				
≤45 years	93	49.2	n.a.	
>45 years	96	50.8	n.a.	
Migration background				
Swiss	173	91.5	n.a.	
Migration background	16	8.5	n.a.	
Language region				
German	132	70.2	78	66.7
French	56	29.8	39	33.3
Education				
Primary	101	54.3	n.a.	
Secondary	62	33.3	n.a.	
Tertiary	23	12.4	n.a.	
Employment				
Employed	150	79.4	n.a.	
Unemployed	39	20.6	n.a.	
Clinical characteristics of the child				
Diagnosis				
Leukemias	74	39.2	46	39.3
Lymphomas	16	8.5	10	8.5
CNS tumors	34	18.0	23	19.7
Neuroblastoma	13	6.9	8	6.8
Retinoblastoma	13	6.9	5	4.3
Renal tumors	12	6.3	8	6.8
Hepatic tumors	4	2.1	3	2.6
Malignant tumors	2	1.1	3	2.6
Soft tissue sarcomas	14	7.4	3	2.6
Germ cell tumors	2	1.1	3	2.6
LCH	2	1.1	3	2.6
Other[a]	3	1.6	0	0.0
Treatment received[b]				
Surgery only	30	16.0	20	17.5
Chemotherapy	118	63.1	74	64.9
Radiotherapy	30	16.0	17	14.9
SCT	9	4.9	3	2.6
Type of treating hospital				
University hospital	160	84.7	102	87.2
Regional hospital	29	15.3	15	12.8

Table 1 contd.

	Participating parents		Non-participating parents	
	N	**%**	**N**	**%**
Total	**189**	**100**	**117**	**100**
Child's age at diagnosis				
0-1 years	58	30.7	35	29.9
2-4 years	82	43.4	48	41.0
5+ years	49	25.9	34	29.1
Time since diagnosis				
5-9 years	64	33.9	38	32.5
10-14 years	96	50.8	58	49.6
15-17 years	29	15.3	21	17.9
Child's age at study				
<14 years	60	31.8	36	30.8
14-15 years	43	22.7	17	14.5
>15 years	86	45.5	64	54.7
Relapse				
No	168	88.9	104	88.9
Yes	21	11.1	13	11.1
Parent-reported late effects				
No	100	54.4	68	64.2
Yes	84	45.6	38	35.8
Parental involvement in follow-up				
No	10	7.1	n.a.	
Yes	130	92.9	n.a.	
Follow-up attendance				
Yes	141	74.6	n.a.	
No	48	25.4	n.a.	
	Participants		**Non-participants**[a]	
	Mean	**SD**	**Mean**	**SD**
Parent's age	46.1	4.8	n.a.	n.a.
Child's age at study	14.7	1.8	15.0	1.9
Child's age at diagnosis	3.4	2.2	3.6	2.4
Time since diagnosis	11.3	2.5	11.4	2.5

Note: Percentages are based upon available data for each variable. Abbreviations: CNS, Central Nervous System; LCH, Langerhans Cell Histiocytosis; n.a., not available; N, Number; SCT, Stem Cell Transplantation; SD, Standard Deviation; [a]Other: malignant epithelial neoplasms, malignant melanomas and other or unspecified malignant neoplasms; [b]Chemotherapy may include surgery, radiotherapy may include chemotherapy and/or surgery.

Table 2. Factors associated with preferences for follow-up care by generalists (GP/Telephone-led follow-up) versus specialists (from univariable logistic regression models)

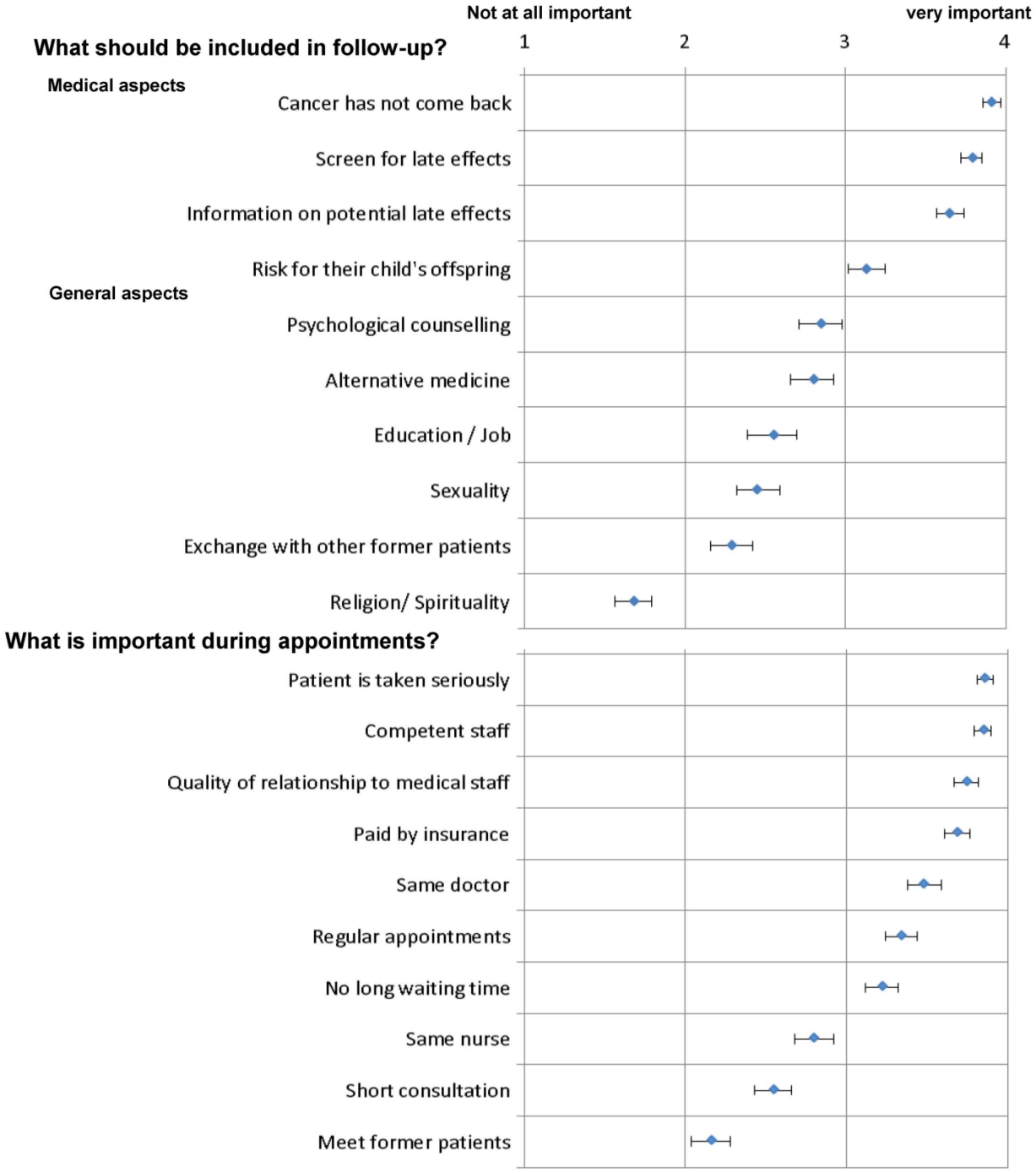
		Preference for generalist follow-up		Univariable regression		
	N Total	N	%	OR	95%CI	p
Socio-demographic characteristics of parents						
Sex						0.300
Female	158	29	18.4	1		
Male	28	3	10.73	0.53	0.15-1.89	
Age at study						0.894
≤45 years	91	16	17.6	1		
>45 years	95	16	16.8	0.95	0.44-2.03	
Migration background						0.176
Swiss	170	31	18.2	1		
Immigrant	16	1	6.3	0.30	0.04-2.35	
Language region						0.127
German	131	19	14.5	1		
French	54	13	24.1	1.86	0.85-4.12	
Education						0.379
Primary	100	20	20.0	1		
Secondary	60	7	11.7	0.53	0.21-1.34	
Tertiary	23	4	17.4	0.84	0.26-2.75	
Employment						0.490
Employed	148	24	16.2	1		
Unemployed	38	8	21.1	1.38	0.56-3.37	
Clinical characteristics of the child						
Diagnosis						0.657
Leukemia	72	16	22.2	1		
Lymphoma	16	3	18.7	0.81	0.20-3.19	
CNS tumor	34	6	17.7	0.75	0.26-2.13	
Neuroblastoma	12	2	16.7	0.70	0.14-3.53	
Bone tumor/STS	16	1	6.3	0.23	0.03-1.90	
Other tumor[a]	24	3	12.5	0.5	0.13-1.89	
Treatment received[b]						0.297
Surgery	29	7	24.1	1		
Chemotherapy	117	19	16.2	0.61	0.23-1.63	
Radiotherapy	29	2	6.9	0.23	0.04-1.24	
SCT	9	2	22.2	0.90	0.15-5.36	
Type of treating hospital						0.080
University hospital	157	30	19.1	1		
Regional hospital	29	2	6.9	0.31	0.07-1.39	
Child's age at diagnosis						0.214
0-1 years	56	6	10.7	1		
2-4 years	82	18	21.9	2.34	0.87-6.34	
5+ years	48	8	16.7	1.67	0.53-5.20	
Child's age at study						0.381
<14 years	60	8	13.3	1		
14-15 years	42	6	14.3	1.08	0.35-3.39	
>15 years	84	18	21.4	1.77	0.71-4.40	
Time since diagnosis						0.556
5-9 years	64	11	17.2	1		
10-14 years	94	18	19.2	1.14	0.50-2.62	
15-17 years	28	3	10.7	0.58	0.15-0.40	

Table 2 contd.

	N Total	N	%	OR	95%CI	p
Relapse						0.083
No	166	31	18.7	1		
Yes	20	1	5.0	0.23	0.03-1.78	
Parent-reported late effects						0.521
No	99	18	18.2	1		
Yes	82	12	14.6	0.77	0.35-1.72	
Parental involvement in follow-up						0.766
No	10	1	10.0	1		
Yes	129	17	13.2	1.36	0.16-11.47	
Concerns about consequences of cancer						0.289
No	50	12	24.0	1		
Medium	54	7	13.0	0.47	0.17-1.31	
High	80	12	15.0	0.56	0.23-1.37	
Follow-up attendance						**0.009**
Yes	140	18	12.3	1		
No	46	14	20.0	2.97	1.33-6.60	
Doctors involved in current care						**0.007**
Specialist	123	13	10.6	1		
Generalist	29	16	33.3	4.23	1.84-9.71	

Note: Numbers for each outcome vary because not all participants answered each question. Percentages are based upon available data for each variable. Abbreviations: CI, Confidence Interval; CNS, Central Nervous System; GP, General practitioner; OR, Odds Ratio; N, Number; p, p-value; SCT, Stem Cell Transplantation; [a]Other: malignant epithelial neoplasms, malignant melanomas and other or unspecified malignant neoplasms; [b]Chemotherapy may include surgery, radiotherapy may include chemotherapy and/or surgery;

Figure 1. Parents' preferences for the organization of follow-up care



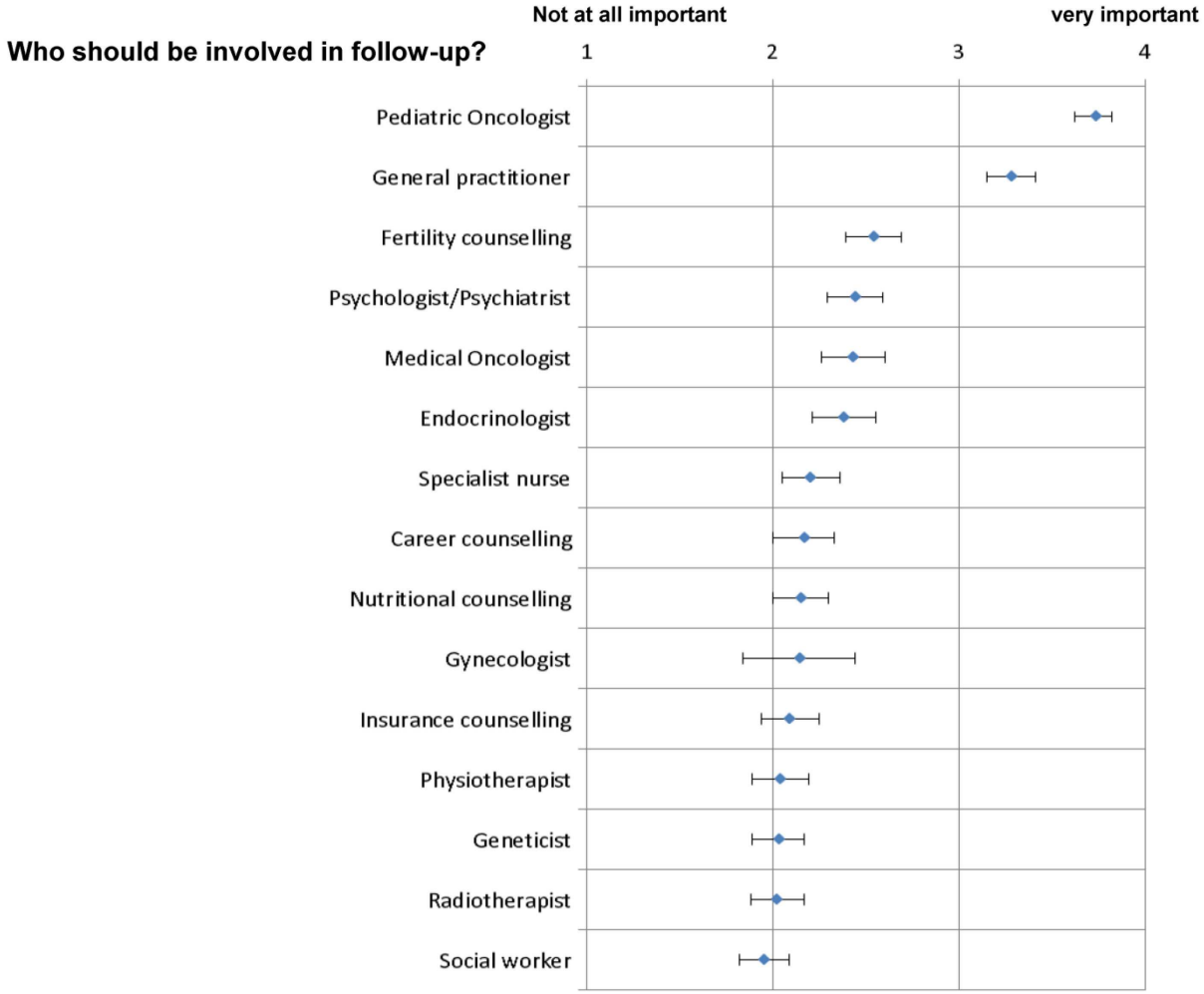

Who should be involved in follow-up?
Not at all important
very important
1
2
3
4
Pediatric Oncologist
General practitioner
Fertility counselling
Psychologist/Psychiatrist
Medical Oncologist
Endocrinologist
Specialist nurse
Career counselling
Nutritional counselling
Gynecologist
Insurance counselling
Physiotherapist
Geneticist
Radiotherapist
Social worker

Figure 2. Parents' preferred model of follow-up care

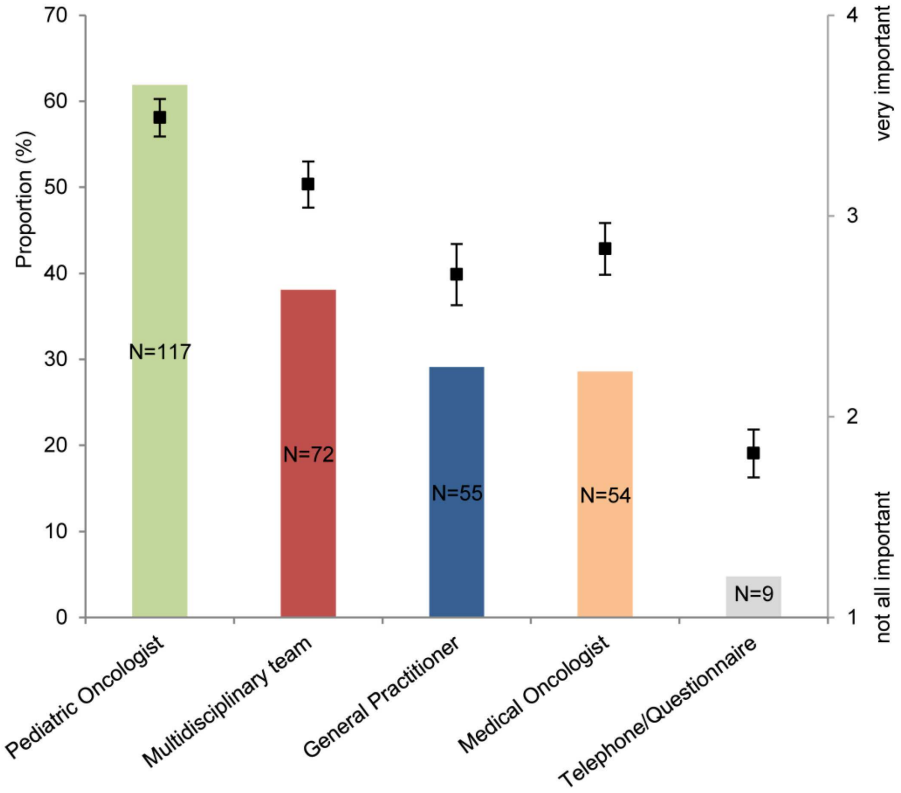


Figure 2 shows on the left side the proportion (bars) of parents rating the respective model as their most preferred follow-up care model (highest mean score among all the models; parents could have more than one preferred model reaching the same highest score), and on the right side the four point likert scale and the mean values (square symbols)
Abbreviations: N=number

Figure 3. Parents' preferred follow-up care model stratified by the model their child is currently attending (Specialist vs generalist)

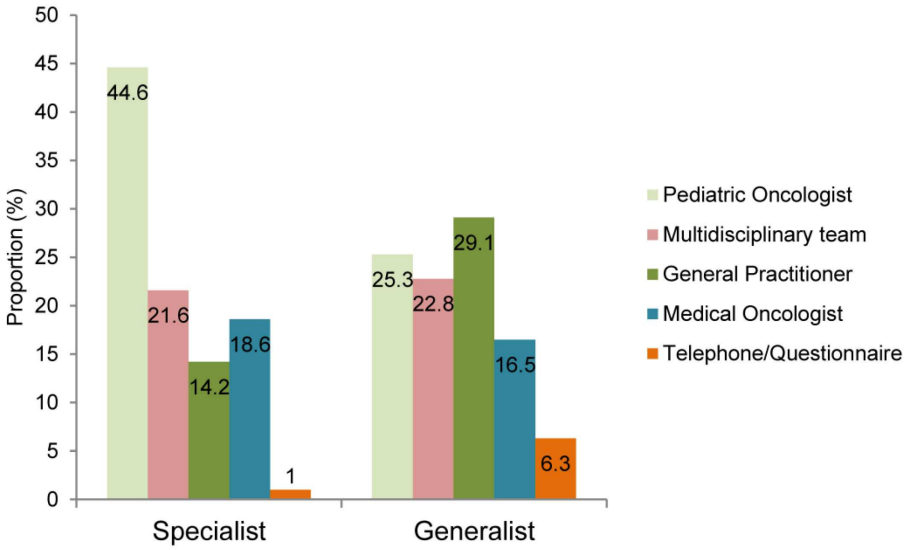


Figure 3 shows the proportion of parents rating the respective model as their preferred follow-up care model stratified by the model their child is currently attending (highest mean score among all the models; parents could have more model which they preferred most)